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Case Report

Central giant cell granuloma of the mandibular condyle: a case report, literature review, and discussion of treatment

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ABSTRACT

Benign and malignant neoplasms of the temporomandibular joint are uncommon. Their presence poses a diagnostic and therapeutic challenge for clinicians. The central giant cell granuloma is a relatively common lesion of the jaws; however, it has been reported rarely to originate from the mandibular condyle. To date, only 5 such cases have been documented. We report a case of a large central giant cell granuloma of the condylar head with extension into the infratemporal fossa in a 29-year-old male. The patient was treated with resection and reconstruction using a costochondral graft. The signs, symptoms, and radiographic features are described and compared with the previous reports in the literature. The therapeutic options detailed in the literature have been focused mainly on lesions occupying the dentate regions of the maxilla and mandible. As such, we will review the surgical and pharmacologic options available to the surgeon and discuss their appropriateness with regard to this unique presentation of the central giant cell granuloma.

1. Introduction

Neoplasms of the temporomandibular joint (TMJ) are uncommon and often confused in the early stages for the more common TMJ dysfunction or derangement [1]. The presence of a central giant cell granuloma (CGCG) in the mandibular condyle has been reported rarely. The purpose of this study is to provide clinicians with a description of the clinical features of a CGCG presenting in the mandibular condyle as well as an analysis of the literature and discussion of the therapeutic strategies available and appropriate for this unique location.

2. Presentation of case

A 29-year-old healthy Caucasian male with a history of a painless left preauricular mass of 6 months' duration was referred by his dentist to the Department of Oral and Maxillofacial Surgery at the Montreal General Hospital for evaluation (Figure 1). A panoramic radiograph taken by the patient's dentist demonstrated a large radiolucent mass of the left condylar process (Figure 2).

The patient noted a progressive enlargement of the mass since its onset and in the days preceding the consultation a new finding of hypoesthesia over the distribution of the mandibular division of the trigeminal nerve while sleeping on his left side. The patient reported no changes in his occlusion but some resistance and mild discomfort on maximal opening. On physical examination, the patient was found to have a firm, nontender, palpable mass in the left preauricular region. Clicking or crepitus of the TMJs bilaterally was absent. The maximal mouth opening, left and right lateral excursive movements were found to be 38 mm, 5 mm, and 5 mm, respectively. There was no deviation of the mandible on opening or closing movements. Trigeminal nerve function was normal when no pressure was applied to the left preauricular region. With prolonged pressure to the left preauricular area, the patient reported hypoesthesia over the distribution of the mandibular nerve.

The computed tomography scan of the facial bones demonstrated a 5.5 × 3.8 × 3.4-cm radiolucent mass emanating from the left condylar head (Figure 3). The condyle was displaced inferiorly in the fossa to accommodate the mass. The outline demonstrated a thin cortex with a few breaks at the periphery. There were no signs of invasion of the surrounding soft tissues. The radiographic appearance of a well-defined, corticated mass without soft tissue invasion or skull base resorption was suggestive of a benign aggressive tumor.

An open biopsy of the left condylar mass was performed under general anesthesia. An intraoperative frozen section analysis favored

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Figure 1. Frontal photograph showing a left preauricular mass.

a benign entity but was inconclusive. The final pathology was consistent with a CGCG (Figure 4). Intact parathyroid hormone (4.00 pmol/L) and total calcium (2.42 mmol/L) studies were determined to be within normal limits, ruling out a brown tumor of hyperparathyroidism.

A decision was made to resect the condylar mass and reconstruct the defect with a costochondral graft under general anesthesia. Given the extension of the tumor into the ramus of the mandible, infratemporal fossa, and medially toward the pterygoid plates, 2 incisions were employed for resection: a preauricular and a trans-oral incision. First, a modified preauricular incision [2] was completed, with dissection down to the lateral aspect of the tumor. Once the latero-anterior and posterior aspects were exposed, the left zygomatic arch was pre-plated then osteotomized to facilitate access for superior dissection into the infratemporal fossa. Access to the medial aspect of the tumor was facilitated by a trans-oral mandibular vestibule incision, allowing for gentle dissection up to the pterygoid plates. Once circumferential dissection was complete, the inferior mandibular resection margin was osteotomized, thus allowing for delivery of the tumor in an inferolateral direction in one segment with the articular disc attached (Figure 5). Access through the trans-oral and preauricular sites facilitated complete and safe dissection of the tumor, while a submandibular incision was used to allow access and placement of the costochondral graft into the proper anatomic site.

Figure 2. Panoramic radiograph displaying a thinly corticated radiolucent lesion of the left mandibular condyle.

The final histopathologic diagnosis was a CGCG with some areas of aneurysmal bone cyst-like features. At the 12-month follow-up appointment, the patient's maximal mouth opening and left and right lateral excursive movements had increased to 45 mm, 8 mm, and 8 mm, respectively, while his preoperative occlusion remained intact.

3. Discussion

To date, 5 cases of CGCGs centering on the mandibular condyle have been published in the English literature (Table 1) [3–7].

Table 1
Comparison of published cases of CGCGs originating in the mandibular condyle.

	Shensa and Nasseri [3]	Tasanen et al. [4]	Abu-El-Naaj et al. [5]	Jadu et al. [6]	Munzenmayer et al. [7]	Current study
Age at presentation	15	59	15	31	19	29
Gender	Male	Male	Female	Male	Female	Male
Clinical findings	Expansile mass	Firm, preauricular swelling; 20-mm mouth opening with deviation to ipsilateral side	Preauricular swelling; no restriction in mandibular movements or occlusal changes	Firm, nodular, preauricular swelling	Examination was normal; incidental radiographic finding	Firm, nodular, preauricular swelling; nontender; no change in occlusion
Imaging features	Well-defined radiolucency	Well-defined, multilocular radiolucency	3 × 2-cm well-defined unilocular radiolucency with cortical expansion	Well-defined, multilocular radiolucency with granular bone pattern	Well-defined, multilocular radiolucency with granular bone pattern	Thinly corticated, 5.5 × 3.8 × 3.4-cm multilocular mass
Symptoms	Asymptomatic	Noted painless, slow growing preauricular lump	Asymptomatic	Dull aching pain and progressive limitation in mouth opening over 2 y	Asymptomatic	Mild discomfort and resistance on maximal opening; mandibular nerve hypoesthesia when sleeping on ipsilateral side
Management	Enucleation	Resection and reconstruction with costochondral graft	Enucleation	Enucleation	Resection and reconstruction with nonvascularized fibula graft	Resection and reconstruction with costochondral graft
Outcome	N/A	No evidence of recurrence at 21 mo	No evidence of recurrence at 6 mo	Recurrence requiring resection and alloplastic total joint replacement	No evidence of recurrence at 24 mo	No evidence of recurrence at 6 mo

N/A, not applicable.

Figure 3. CT scan, soft tissue window, demonstrating a 5.5 × 3.8 × 3.4-cm radiolucent mass of the left mandibular condyle in the (A) axial (B) coronal, and (C) sagittal planes.

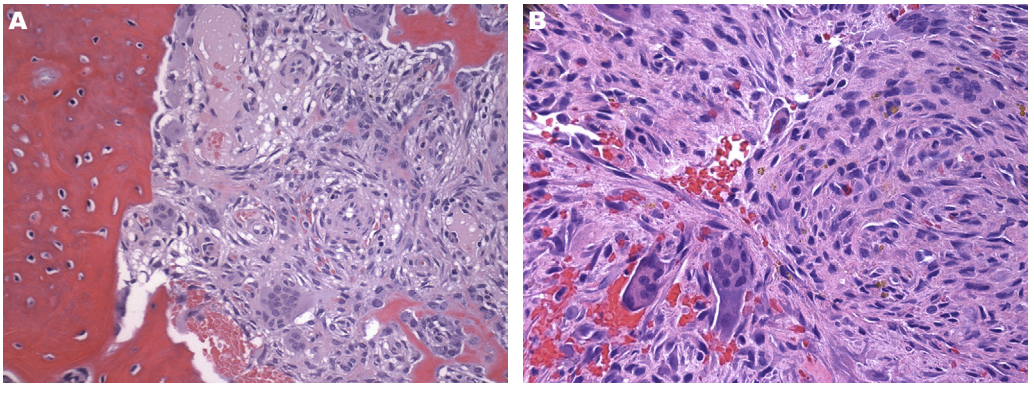

Figure 4. H&E stained photomicrographs (A) 50× and (B) 400× showing a cellular matrix predominantly of oval and spindle-shaped stromal cells interspersed with multinucleated giant cells. There is extensive extravasation of red blood cells and hemosiderin deposition (B).

Including our case report, there is a 2:1 (M-F) gender predilection. This is in contrast to most previous publications that indicate a female tendency. The mean age at presentation is 28 years of age (range 15-59). Of the 6 patients, 4 were asymptomatic on presentation. The remaining 2 patients experienced mild discomfort presumably related to the mass effect of the tumor and not inherent to the neoplasm itself. Three patients presented with a mild restriction in mouth opening and all patients reported no changes in occlusion. Clinically, 5 of the 6 patients developed firm preauricular swellings. Radiographically, the lesions were predominately multilocular in nature and surrounded by a well-defined cortical layer.

The management of the CGCG is controversial. Multiple medical management strategies have been proposed over time with varying degrees of success despite not necessarily targeting the now proposed proliferative cell (spindle-shaped mononuclear stromal cell) [8]. A 2009 Cochrane Review concluded that there was only 1 randomized controlled trial available and no high-quality evidence to support the use of nonsurgical therapies in the management of CGCGs [9].

Surgical management of the CGCG is still the most common treatment modality employed. Broadly, it can be broken into 2 major interventions: curettage ± adjunctive treatments (eg cryotherapy, peripheral ostectomy) and resection. With curettage recurrence rates from 11% to 49% [10,11] have been reported. A study by de Lange and Van den Akker [12] followed up 80 patients after curettage. Patients with aggressive lesions had a recurrence rate of 37.5% while

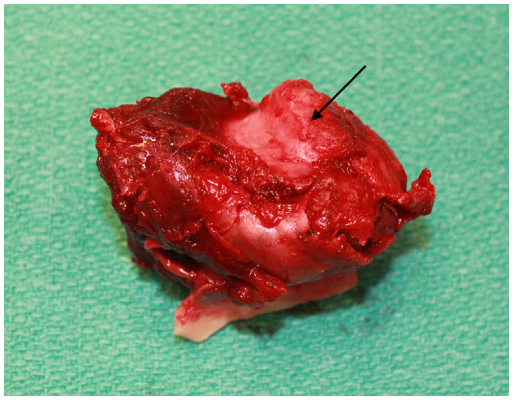

Figure 5. The surgical specimen demonstrating a thinly corticated mass with multiple small perforations. Articular disc is attached superiorly (arrow).

patients classified as having nonaggressive lesions had a recurrence rate of 23.4%. Bataineh et al. [13] evaluated the effectiveness of resection with a 1-cm margin in 18 patients with aggressive CGCGs of the mandible. During the follow-up period only one patient recurred (6.8%). Disease control is superior with resection; however, it is also more frequently associated with tooth loss, damage to developing tooth germs, neurosensory disturbances, fracture, and the use of general anesthesia.

In 1988, Jacoway et al. [14] described a protocol that involved intralesional injection of equal parts of triamcinolone hexacetonide and 0.5% bupivacaine (1:200,000 epinephrine). Ten milliliters of solution was injected per 1 cm of lesion, as determined on Panorex imaging, weekly over a period of 6 weeks. The original rationale for this therapy was the histologic resemblance of the CGCG to sarcoid lesions, which were known to respond to steroids [15]. The largest case series to date was published by Nogueira et al. [16]. Of the 21 patients in the study, 15 showed a good response, 4 demonstrated a moderate response, and 2 patients had a negative response. Furthermore, 8 of 19 patients with a moderate-to-good response (increased radiopacity) still required an osteoplasty to reestablish facial aesthetics.

Harris [17] was the first to propose the use of subcutaneous human calcitonin for the treatment of CGCGs. Due to its commercial availability, salmon calcitonin is now used either subcutaneously or intranasally. In 2003, Pogrel [15] published a series of 10 cases. Nine patients received daily subcutaneous injections while 1 patient elected for the intranasal route of administration. In all cases, there were no radiographic changes at 4-6 months, but thereafter calcification appeared quickly up to 18 months. Of the 9 patients receiving subcutaneous injections one recurred after 26 months and was curettage was performed, while the patient receiving the intranasal calcitonin abandoned the treatment after 4 months and curettage was performed as well. Treatment durations ranged from 19 to 26 months in the 7 patients free of recurrence. Similarly to corticosteroid treatment, the use of calcitonin may result in an increase in radiopacity and histologic resolution of the lesion, but may not necessarily restore the normal bony architecture of the jaw.

Kaban et al. [18] were the first to describe the successful use of interferon alfa-2a in the treatment of an aggressive CGCG. Among its other effects, interferon alfa-2a is a known inhibitor of basic fibroblast growth factor. At this time, proliferative vascular lesions, namely, the hemangioma, known to overexpress basic fibroblast growth factor, had been treated successfully with interferon alfa-2a. It is now appreciated that the CGCG likely does not represent a proliferative vascular lesion; however, the angiogenic factors may play a role in the osteoclastogenesis and thus in the growth of the lesion. Kaban et al. [19] further demonstrated the efficacy of interferon alfa-2a in a retrospective case series of 16 patients with aggressive CGCGs. All patients were initially treated with curettage followed by interferon therapy for a mean duration of 7 months. No tumor recurrence was seen in this population after at least 2 years of therapy. In this series 88.5% of patients developed 1 or more side effects, 42.3% required their dose to be reduced as a result, and 15.3% developed side effects necessitating cessation of treatment. The role of interferon alfa-2a as a monotherapy is less certain as it does not target the suspected proliferative spindle-shaped stromal cell.

When managing neoplasms of the mandibular condyle, nonsurgical management with preservation of the native condyle is highly desirable. Unfortunately, there is no medical management strategy that appears to be effective in every individual patient with a CGCG. Based on the literature, condylar resection with immediate reconstruction has the lowest recurrence rate with the shortest duration of treatment. Interferon alfa-2a therapy has demonstrated the best results when used as adjuvant therapy following curettage. The morbidity associated with curettage of a large condylar lesion and resection is similar. As such, this treatment strategy offers little advantage over resection, given the disadvantages of prolonged treatment duration and high incidence of side effects. Intralesional corticosteroid treatment requires multiple injections over a period of at least 6 weeks [14]. Repeated injections in the TMJ and infratemporal fossa regions are not without risk of neurovascular injury possibly resulting in paresthesia, paresis, hematoma, or pseudoaneurysm formation. Additionally, the administration of solution into the deep aspect of the lesion would be challenging, possibly compromising the outcome. The administration of subcutaneous calcitonin-like corticosteroids has had some success but requires daily subcutaneous injection and an extended treatment duration often spanning over 2 years [15]. Unfortunately, the intranasal form has demonstrated limited success thus far [20]. The side effects of calcitonin are usually minimal but can include nausea, flushing, and dizziness [20]. In medical treatment of large lesions, the normal anatomic form is often not achieved [15,16] and further osteoplasty is often required for a desirable cosmetic result. Achieving a normal anatomic form of the TMJ is of paramount importance, further complicating medical management strategies. A goal less than this will result in residual facial deformity and persistence of preoperative symptoms that may include limitation in mouth opening, pain, and neurosensory disturbances. Based on the available literature, it was determined that condylar resection with immediate reconstruction would be the most appropriate treatment for a large, aggressive, symptomatic, and deforming CGCG of the mandibular condyle in the adult patient. The role of nonsurgical therapies may be increasingly important when condylar CGCGs are asymptomatic, small, and classified as nonaggressive in the growing individual.

Conflicts of interest

The authors declare there are no conflicts of interest.

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References

[1] Poveda-Roda R, Bagan J, Sanchis J, Margaix M. Pseudotumors and tumors of the temporomandibular joint. A review. Med Oral Patol Oral Cir Bucal 2013;18:392–402.

[2] Al-Kayat A, Bramley P. A modified pre-auricular approach to the temporomandibular joint and malar arch. Br J Oral Maxillofac Surg 1979;17:91–103.
[3] Shensa DR, Nasseri S. Central giant cell reparative granuloma of the mandibular condyle. J Oral Surg 1978;36:642–3.
[4] Tasanen A, von Konow L, Nordling S. Central giant-cell lesion in the mandibular condyle. Report of a case. Oral Surg Oral Med Oral Pathol 1978;45:532–9.
[5] Abu-El-Naaj I, Ardekian L, Liberman R, Peled M. Central giant cell granuloma of the mandibular condyle: a rare presentation. J Oral Maxillofac Surg 2002;60:939–41.
[6] Jadu FM, Pharoah MJ, Lee L, Baker GI, Allidina A. Central giant cell granuloma of the mandibular condyle: a case report and review of the literature. Dentomaxillofac Radiol 2011;40:60–4.
[7] Munzenmayer J, Tapia P, Zeballos J, Martínez A, Compan Á, Urra A, et al. Central giant cell granuloma of the mandibular condyle: case-report. Rev Clin Periodoncia Implantol Rehabil Oral 2013;6:83–6.
[8] Liu B, Yu S-FF, Li T-JJ. Multinucleated giant cells in various forms of giant cell containing lesions of the jaws express features of osteoclasts. J Oral Pathol Med 2003;32:367–75.
[9] Suárez-Roa MDL, Reveiz L, Ruiz-Godoy Rivera LM, Asbun-Bojalil J, Davila-Serapio JE, Menjivar Rubio AH, et al. Interventions for central giant cell granuloma of the jaws. Cochrane Database Syst Rev 2009;(4):CD007404.
[10] Whitaker SB, Waldron C. Central giant cell lesions of the jaws. Oral Surg Oral Med Oral Pathol 1993;75:199–208.
[11] Eisenbud L, Stern M, Rothberg M, Sachs SA. Central giant cell granuloma of the jaws: experiences in the management of thirty-seven cases. J Oral Maxillofac Surg 1988;46:376–84.
[12] de Lange J, Van den Akker HP. Clinical and radiological features of central giant-cell lesions of the jaw. Oral Surg Oral Med Oral Pathol 2005;99:464–70.
[13] Bataineh AB, Al-Khateeb T, Rawashdeh MA. The surgical treatment of central giant cell granuloma of the mandible. J Oral Maxillofac Surg 2002;60:756–61.
[14] Jacoway JR, Howell FV, Terry BC. Central giant cell granuloma: an alternative to surgical therapy. Oral Surg Oral Med Oral Pathol 1988;66:572.
[15] Pogrel MA. Calcitonin therapy for central giant cell granuloma. J Oral Maxillofac Surg 2003;61:649–53.
[16] Nogueira RL, Teixeira RC, Cavalcante RB, Ribeiro RA, Rabenhosrt SH. Intralesional injection of triamcinolone hexacetonide as an alternative treatment for central giant-cell granuloma in 21 cases. Int J Oral Maxillofac Surg 2010;39:1204–10.
[17] Harris M. Central giant cell granulomas of the jaws regress with calcitonin therapy. Br J Oral Maxillofac Surg 1993;31:89–94.
[18] Kaban LB, Mulliken JB, Ezekowitz A, Ebb PD, Smith PS, Folkman J. Antiangiogenic therapy of a recurrent giant cell tumor of the mandible with interferon alfa-2a. Pediatrics 1999;103:1145–9.
[19] Kaban LB, Troulis MJ, Wilkinson MS, Wilkinson MJ, Ebb D, Dodson TB. Adjuvant antiangiogenic therapy for giant cell tumors of the jaws. J Oral Maxillofac Surg 2007;65:2018–24.
[20] de Lange J, van den Akker HP, Veldhuijzen van Zanten GO, Engelshove HA, van den Berg H, Klip H. Calcitonin therapy in central giant cell granuloma of the jaw: a randomized double-blind placebo-controlled study. Int J Oral Maxillofac Surg 2006;35:791–5.